Spinal Cord Injury and Spinal Fracture in Patients with Ankylosing Spondylitis

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Research article

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Abstract

Background

Spinal cord injury (SCI) and spinal fracture are major complications in patients with ankylosing spondylitis (AS) who sustain spinal trauma. The purpose of this study is to investigate the incidence, predictors, and sequelae of spinal trauma in patients with AS.

Methods

This study evaluated our AS patients who had spinal traumas between January 1, 2006, and June 30, 2016 and a comparison of those patients with SCI alone, fracture alone and both SCI and spinal fracture.

Results

105 patients were enrolled. Of these patients, 89.5% had spinal fractures, and 57.1% had SCI. Among the patients with spinal fractures, 52.1% had SCI. The existence of bamboo spine was significantly more frequent in the fracture group (78.7% vs. 36.4%; $P = 0.006$) than in the non-fracture group. The SCI patients had more subluxation or dislocation (48.3% vs. 8.9%; $P < 0.001$), and more cases of spinal epidural hematoma (SEH) (21.7% vs. 2.2%; $P = 0.003$) than the non-SCI patients. The rate of delayed diagnosis for spinal fracture was 31.4%, where 1/3 of these patients developed delayed SCI. Among the incomplete SCI patients, 58.3% had neurological improvement after treatment ($P = 0.004$).

Conclusions

The patients with existing bamboo spine at X-ray had a higher spinal fracture rate. Spinal fractures involving the C3-C7 region, subluxation or dislocation, spinal fracture severity, and a SEH were found to be predictive of SCI. SCI in AS patients resulted in a high mortality and complication rates.

Introduction

Ankylosing spondylitis (AS) is a chronic inflammatory disease mainly affecting the axial skeleton that is characterized by progressive bone loss, erosion, and syndesmophyte formation, leading to progressive rigidity and altered biomechanical properties of the spine.[1] AS thus increases the risk of vertebral fractures, even from minor injuries.[2–4] Often, AS patients have marginal syndesmophyte formation, which presents as the classic “bamboo spine” appearance in radiographic examinations.[5] The ankylosed spine is very fragile due to secondary osteoporosis and loss of mobility.[6] The risk of fracture is even higher when the disease develops for a longer period of time. The progressively ankylosed spine is more susceptible to injury[2,7,8] and relatively unstable compared to those in a normal spine.[9] Otherwise, the delayed diagnosis of spinal fractures in AS patients after minor trauma is common. Patients with delays in diagnosis often present with chronic pain, progressive neurologic deficits, and worsening spinal deformity.[10–12]

Spinal trauma includes spinal cord injury (SCI), spinal fracture, or both. Spinal trauma in the ankylosed spine has a large impact on morbidity and mortality.[3,11,13] The reported prevalence of spinal fracture in patients with AS varies widely between 10% and 17%,[6,8,14,15] and the rate of SCI ranges from 19–91%.[4,11,14,16,17] Because of uncommon disease entity, the reported information about the predictors of these conditions remains scarce.[4,13] The purpose of this study is to investigate the incidence, predictors, and sequelae of spinal trauma in AS patients in a single tertiary center.

Material And Methods

Patient sample

6285 spinal fractures or spinal cord injury after spinal injury patients were treated from January 1, 2006 to June 30, 2016 in our institution, only 110 AS patients with spinal trauma were identified. The patients' charts and images were reviewed by 3 experienced neurosurgeons. The diagnosis of AS was reconfirmed by rheumatologist according to the Assessment of Spondyloarthritis International Society (ASAS).[18-21] 105 patients with at least 2-year follow-up or died during follow-up duration were included. 4 patients followed less than 1 year and 1 patient with loss of key image data were excluded. This study was approved by the Institutional Review Board (IRB No.: 201700858B0).

Variables

Predefined and generally accepted parameters (listed in Table 1) were extracted from the electronic medical records, including patient's sex and age at time of injury, initial neurologic grading with the American Spinal Injury Association (ASIA) Impairment Scale (AIS), presence of high-energy trauma,[22] presence of bamboo spine, presence of subluxation or dislocation, spinal cord injury and level, spinal fracture and level, spinal epidural hematoma(SEH) and level, fracture classification, treatment, outcome at discharge and two years after trauma, and complications.

Outcome measures

For each patient, we manually graded the degree of SCI using the AIS at the time of the initial SCI diagnosis, at the time of discharge, and at the 2-year follow-up. Patients with an AIS grade decline of 1 or more (such as a decline from E to D) at any point after injury were considered SCI patients. We categorized SCI as either complete (AIS Grade A) or incomplete (AIS Grades B–D). Patients with SCI who showed an improvement of at least 1 AIS grade (such as from D to E) during follow-up were considered neurologically improved.

Statistical methods

Data were presented as frequency and percentage for continuous variables or mean and standard deviation for categorical variables. The investigated patients were divided into three study groups: those with both SCI and fracture, those with SCI alone, and those with fracture alone. The patient characteristics among the study groups were compared by one-way analysis of variance for continuous variables or by Fisher's exact test for categorical variables. The AIS grade improvement from first symptoms to the last follow-up in the patients with SCI was tested using the McNemar test. All tests were 2-tailed and $P < 0.05$ was considered statistically significant. No adjustment of multiple testing (multiplicity) was made in this study. Data analyses were conducted using SPSS 25 (IBM SPSS Inc., Chicago, Illinois).

Results

Of those 105 patients, 49 (46.67%) who suffered from both SCI and spinal fracture, 11 (10.48%) had SCI alone, and 45 (42.85%) had spinal fracture alone. Only a few of the patients were female (4.8%; 5/105). Fifty-three patients (50.5%) suffered from only low-energy trauma. Thirty-three patients (31.4%) had subluxation or dislocation. Ninety-four patients (89.5%) had spinal fractures. The spinal fracture involving the C3 to C7 region was the most frequently in patients with both SCI and spinal fracture (65.3%). The thoracic spine, meanwhile, was the most frequently fractured region in the non-SCI patients (44.4%). Out of all 105 patients, 60 (57.1%) had SCI, including 12 with complete SCI, of which C3-7 spinal cord injury accounted for 43.8%. Forty-nine patients (52.1%) had both SCI and spinal fracture. Seventy-eight patients (74.3%) presented with "Bamboo spine" in X-ray images (Table 2).

Thirty-three patients (31.4%) had delayed diagnosis. Among these patients, all had spinal fracture; 6 of those patients were categorized in the "doctor delay" group, with 2 of those patients subsequently developing SCI at 5 days and 15 days after the trauma, respectively. The remaining 27 patients were categorized in the "patient delay" group, 9 of them developed delayed SCI within 2 to 90 days after the trauma. The average time between the trauma and the confirmed diagnosis for all the patients was 19.8 days (SD = 20.4 days), with a range from 2 to 90 days (Table 2).

32 patients received surgical treatment for spinal fracture only, at 13 ± 93.7 days after trauma, and 45 patients received surgical treatment for SCI consisting of decompression, open reduction, and fixation at 1.8 ± 3.2 days. 6 patients with SCI and spinal fracture, including 3 patients with AIS grade A, received close reduction and halo-jacket fixation and other 3 patients with mild SCI (initial AIS grade: D) with mild spinal fracture (AO spine fracture classification: A) received conservative treatment. 13 patients with mild spinal fractures (AO spine fracture classification: A) without SCI received conservative treatment. Ten of 11 patients with SCI alone were central cord syndrome and recovery to AIS grade E; and the other one was severe head injury without recovering to clear. Their injury levels were one patient in C3, four in C4, two in C5, three in C6 and one in C7. Two of them received anterior microdisectomy with interbody fusion and fixation, the other 8 patients with mild SCI (initial AIS grade: D), and 1 who initially presented a deep coma received conservative treatment.

Fourteen patients (13.3%) had SEH. During follow-up, 32 patients (30.5%) had complications, with infection being the most commonly occurring complication (22.9%). Four patients (3.8%) died during the period of 2-year follow-up, 3 due to pneumonia-related septic shock, and 1 due to pulmonary embolism and cardiac failure with acute respiratory distress syndrome (Table 2).

Among the SCI patients, the AIS grade was significantly improved from the time of the first symptoms to the follow-up after 2 years ($P = 0.004$). Among all 60 patients with SCI, 35 experienced an AIS grade improvement, but 4 patients had worse AIS grades due to the patients who died (n = 4) being given an AIS grade A at the 2-year follow-up (Table 3). Excluding mild SCI (initial AIS grade: D) patients without surgery, 53.3% of 45 patients received surgical treatment got the benefit of long-term neurological recovery.

Comparing the patients with and without spinal fracture, the presence of bamboo spine was significantly more frequent in the fracture group (78.7% vs. 36.4%; $P = 0.006$). The demographic and clinical characteristics were not significantly different, however, between the fracture and non-fracture groups or between the SCI and non-SCI groups. However, the results showed that the SCI patients had more cases involving subluxation or dislocation (48.3% vs. 8.9%; $P < 0.001$), spinal fractures involving the C3-C7 region (53.3% vs. 11.1%; $P < 0.001$), the presence of spinal fracture in general ($P < 0.001$), and the presence of SEH (21.7% vs. 2.2%; $P = 0.003$) than the non-SCI patients (Table 4).

Discussion

Among AS patients, SCI is a major complication regardless of spinal fracture or not. Our results showed that the SCI rate after spinal trauma in AS patients was 57.1% (60/105), while the SCI rate in cases also involving spinal fractures was 52.1% (49/94), which was lower than the average 67.2% rate reported in the largest meta-analysis,[4] but higher than the 32.1% rate reported in the largest single-institution series.[23] Lukasiewicz et al. reported a 21.2% rate of SCI in patients with AS and spinal fracture.[16] Another two single-institution studies reported 58% and 19.7% rates.[11,13] These inconsistent rates of SCI after spinal fractures in AS patients may be due to differences in medical referral standards, different severities of trauma, and different severities of AS.

The diagnoses of fractures in AS patients are frequently delayed, ranging from 17.1–65.4%[4,11,23] as patients are known to have chronic pain even in the absence of trauma. Therefore, aggravating pain following a minor trauma may be overlooked. Due to alterations in bone density, radiologic assessment for fracture in AS patients could be difficult and easily misinterpreted, especially in cases involving fractures at the thoracic spine and thoracolumbar junction. In our study, the rate of delayed diagnosis for spinal fracture was 31.4% (33/105), with 69.7% (23/33) of those cases involving fractures between the T8 and L1 vertebrae. Six of the 33 cases were attributed to a doctor’s oversight, while the remaining cases were due to the patients’ delayed visits. Overseeing may expose these patients to high risk of delayed SCI. In our study, one-third of these 33 patients with a delayed diagnosis of spinal fracture developed delayed SCI. In the initial post-trauma period, 90.9% of these patients had axial pain such as neck or back pain, whereas 33.3% had limb numbness. The attending physician should be reminded of the highly possible devastating consequences of AS patients, even after a low-energy trauma. We suggest routine radiographic examination for all AS patients after trauma and additional computed tomography (CT) if axial pain progresses. Magnetic resonance imaging is a reasonable option for spinal cord injuries and to rule out occult fractures.[12,24–26]

Bamboo spine is a radiographic feature seen in AS that occurs as a result of vertebral body fusion by marginal syndesmophytes. The consequential radiographic appearance is radiopaque spicules that completely bridge the adjoining vertebral bodies. In our study, 74.3% (78/105) patients exhibited this feature. We also observed that these patients with existing bamboo spine had a higher spinal fracture rate than those without bamboo spine ($p = 0.006$, Table 4). However, the results showed no significant correlation between bamboo spine and SCI ($p = 0.367$, Table 4). This contradicted the finding that AS with spinal fracture was significantly related to SCI ($p < 0.001$). To clarify this, we analyzed the relationship between bamboo spine and spinal fracture in the 60 patients with SCI. Four of the 11 patients who had SCI without spinal fracture presented with bamboo spine, while 43 of the 49 patients with both SCI and spinal fracture had bamboo spine. A comparison of these two groups revealed a significant difference ($p = 0.001$, Table 2). Patients with non-complex compression fractures with the posterior ligamentous complex (PLC) intact (Orthopedic Trauma Association classification type A) had a lower rate of SCI than those with complex fractures, such as flexion/distraction type (B.1 and B.2, PLC disrupted), hyperextension type (B.3), or shear/rotation type (type C) fractures ($p < 0.001$). Subluxation or dislocation was also found to be a risk factor for the AS patients in developing SCI ($p < 0.001$). The severity of spinal structure disruptions is thus predictive of SCI in AS patients. SCI in cases involving a complex fracture is not only caused by the destruction resulting from the direct impact, but also by further compression from bone fragments, hematoma, or disk material.[27] Thus, we hypothesize that AS patients with existing bamboo spine have a high probability of experiencing spinal fracture, but such a mild fracture will not necessarily be severe enough to cause SCI.

There are no universal guidelines for the management of spinal trauma in AS patients.[26] Non-operative treatment has long been recommended for fractures of ankylosed spines.[26,28] The management options include bed rest, skeletal traction, bracing, or immobilization with a halo-vest.[29] Conservative methods are reasonable for non-displaced or minimally displaced fractures.[28] However, under the inherent instability of these fractures and their high potential of acute displacement, catastrophic situation may occur.[30] Therefore, surgical fixation with long segmental instrumentation combined with fusion is highly recommended.[30] Furthermore, the compression of neurologic elements often requires surgical evacuation. Recent studies have demonstrated a trend for higher complication rates in non-operative patients. Patients with bed rest are associated with higher pulmonary complications[29] and present a risk of neurological deterioration.[3,4,29] Surgical stabilization usually includes anterior, posterior, or combined fixation, often accompanied by decompression with laminectomy and various osteotomy techniques for deformity correction.[31,32] In our study, 3 patients with initial AIS A received close reduction and halo-jacket fixation and all 3 patients (100%) had complications: one experienced screw loosening, two developed pneumonia. In contrast, seven of 9 patients (77.8%) with initial AIS A had complication after surgery, suggesting a trend of lower complication rates in severe SCI.

All 14 patients with SEH developed SCI, thus making SEH an important predictive factor of SCI ($p = 0.003$). SEH occurred in 13.3% of the AS patients, which is much higher than in the rates ranged from 0.5–7.5% of the general population.[33,34] As opposed to severe fractures that caused SCI immediately, SEH may bring about subacute SCI hours after the trauma. The mechanisms underpinning cervical SEH formation are not fully understood, but disruption of the posterior longitudinal ligament and spinal epidural vessel rupture may play an important role.[34,35] Symptomatic SEH is thought to be a neurosurgical emergency. In the general population, better long-term neurological recovery was noted after early surgical intervention. However, no major case study has reported in AS patients. In our study, the investigated SCI patients received surgical treatment at 1.8 ± 3.2 days after trauma. Some of these patients had delayed surgical treatment due to delayed diagnosis, old age, higher co-morbidity, or poly-trauma requiring other treatments.

None of the patients who had complete SCI at admission showed improvement in AIS grade after 2 year, as compared to the 58.3% (35/60) of patients in the incomplete SCI group who showed improvement. Overall, AS patients with incomplete SCI had better long-term neurological recovery. Complication rates were significantly higher ($p = 0.001$) in patients with SCI. The 4 patients who died within 1 year were significantly older (with an average age of 69.2 years). This echoes the findings of higher mortality in older AS patients after spinal fracture in previous studies.[11,13] In our study, although these AS patients with the different complexity of fracture with different degree of SCI, more than 53% patients get the benefit of long-term neurological recovery after surgical treatment.

Conclusions

SCI is a major complication of spinal trauma in AS patient. After spinal trauma, AS patients with existing bamboo spine have higher rates of spinal fracture. A cervical fracture involving the C3-C7 region, subluxation or dislocation, higher fracture severity, and SEH are predictive of SCI. For AS patients, even a low-energy trauma could cause a spinal trauma including SCI or fracture. Delayed diagnosis of fracture and SCI happens in about 30% and 10% of cases of spinal trauma. Therefore, attending physicians should be reminded of these potential effects in AS patients.

List Of Abbreviations

SCI
Spinal cord injury; AS:ankylosing spondylitis; SEH:spinal epidural hematoma; ASAS:Assessment of Spondyloarthritis International Society; AIS:American Spinal Injury Association Impairment Scale.

Declarations

Ethics approval and consent to participate: For retrospective studies, formal consent is not required. The database of all patients and procedures was retrospectively reviewed to identify the patients, and the study was approved by our institute's institutional review board (IRB No.: 201700858B0).

Consent for publication: Informed consent was obtained from all individual participants included in the study.

Availability of data and materials: The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Competing interests: All authors certify that there is no actual or potential conflict of interest in relation to this article, and there are no financial interests to disclose.

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Tables

Table 1
Definition of parameters in the article

Low-energy trauma: not high-energy trauma, such as high-speed traffic accidence or fall > 15 feet[13]
Delayed diagnosis (spinal fracture or SCI diagnosis after the day of trauma, less than 24 hours count 0 day) **Patient's delay**: the patient visits a physician after the day of trauma **Doctor's delay**: the patient was not diagnosed by doctor
Bamboo spine: diagnosis by radiologist's report
Subluxation: defined as more than 2 milli-meter distance between the inferior endplate of the neighboring superior vertebra and the superior endplate of the neighboring inferior vertebra at the anterior longitudinal ligament line or dislocation at spinal fracture or spinal cord injury level
Spinal cord injury and level, spinal fracture and level, spinal epidural hematoma and level (levels C0–C2, C3–C7, T1–T12, L1–L5)
Spinal epidural hematoma: spinal epidural hematoma detected on initial or subsequent computed tomography and/or magnetic resonance imaging
Fracture classification **C0–C2** Atlas fractures classified according to Levine and Edwards,[36] fractures of the odontoid process according to Anderson and D'Alonzo,[37] and fractures of the odontoid body according to Levine and Edwards.[38] **C3–L5** Fractures classified according to an algorithm derived from the AO Spine fracture classification.[39]
Discharge outcome, 2 **years after trauma outcome**: AIS
Complications: all events associated with treatment and associated with SCI occurring within 2 years after the trauma. **Treatment associated**: instrumentation failure, such as migration or loosening of screws/rods; wound infection **Spine or spinal cord injury associated**: respiratory failure; pneumonia; pulmonary embolism; pneumothorax; decubitus ulcer; urinary tract infection; sepsis

Table 2
Characteristics in patients with ankylosing spondylitis according to the spinal cord injury with or without fracture

Variables	**Whole cohort (*N* = 105)**	**SCI and fracture (*n* = 49)**	**SCI alone (*n* = 11)**	**Fracture alone (*n* = 45)**	***P* value**
Male	100 (95.2)	48 (98.0)	10 (90.9)	42 (93.3)	0.312
Age (year)	56.2 ± 12.3	56.6 ± 10.2	55.9 ± 6.2	55.9 ± 15.3	0.968
Low-energy trauma	53 (50.5)	23 (46.9)	6 (54.5)	24 (53.3)	0.814
Subluxation or dislocation	33 (31.4)	28 (57.1)	1 (9.1)$_a$	4 (8.9)$_a$	< 0.001
Location of spinal fractures					< 0.001
None	11 (10.5)	0 (0.0)	11 (100.0)$_a$	0 (0.0)$_b$	
C0-2	5 (4.8)	1 (2.0)	0 (0.0)	4 (8.9)	
C3-7	37 (35.2)	32 (65.3)	0 (0.0)$_a$	5 (11.1)$_a$	
T	28 (26.7)	8 (16.3)	0 (0.0)	20 (44.4)$_{ab}$	
L	15 (14.3)	1 (2.0)	0 (0.0)	14 (31.1)$_{ab}$	
Multiple regions involved	9 (8.6)	7 (14.3)	0 (0.0)	2 (4.4)	
Spinal fracture classification					< 0.001
None	11 (10.5)	0 (0.0)	11 (100.0)$_a$	0 (0.0)$_b$	
A1-A3	13 (12.4)	2 (4.1)	0 (0.0)	11 (24.4)$_a$	
B1-B3	42 (40.0)	24 (49.0)	0 (0.0)$_a$	18 (40.0)$_b$	
C1-C3	36 (34.3)	23 (46.9)	0 (0.0)$_a$	13 (28.9)$_b$	
Others	3 (2.9)	0 (0.0)	0 (0.0)	3 (6.7)	
Location of SEH					0.006

SCI, spinal cord injury; SEH, spinal epidural hematoma

Values are given as frequency (%) or mean ± standard deviation

‘a’ and ‘b’ indicate the value reached a significant difference versus the “SCI and fracture” and “SCI alone” groups, respectively

Variables	Whole cohort (*N* = 105)	SCI and fracture (*n* = 49)	SCI alone (*n* = 11)	Fracture alone (*n* = 45)	*P* value
None	91 (86.7)	36 (73.5)	11 (100.0)	44 (97.8)$_a$	
C	8 (7.6)	8 (16.3)	0 (0.0)	0 (0.0)$_a$	
C-T	4 (3.8)	4 (8.2)	0 (0.0)	0 (0.0)	
L, T-L	2 (1.9)	1 (2.0)	0 (0.0)	1 (2.2)	
Bamboo spine	78 (74.3)	43 (87.8)	4 (36.4)$_a$	31 (68.9)$_{ab}$	0.001
Delayed diagnosis	33 (31.4)	12 (24.5)	0 (0.0)	21 (46.7)$_{ab}$	0.003
Delayed diagnosis type (*n* = 33)					1.000
Doctor	6 (18.2)	2 (16.7)	0 (0.0)	4 (19.0)	
Patient	27 (81.8)	10 (83.3)	0 (0.0)	17 (81.0)	
Delayed diagnosis days (*n* = 33)	19.8 ± 20.4	9.1 ± 9.2	-	26.0 ± 22.5	-
SEH	14 (13.3)	13 (26.5)	0 (0.0)	1 (2.2)$_a$	0.001
Complication	32 (30.5)	25 (51.0)	1 (9.1)$_a$	6 (13.3)$_a$	< 0.001
Complication, infection	24 (22.9)	20 (40.8)	1 (9.1)$_a$	3 (6.7)$_a$	< 0.001

SCI, spinal cord injury; SEH, spinal epidural hematoma

Values are given as frequency (%) or mean ± standard deviation

‘a’ and ‘b’ indicate the value reached a significant difference versus the “SCI and fracture” and “SCI alone” groups, respectively

Table 3

AIS grade improvement from first symptoms to the follow-up after 2 years in patients with spinal cord injury and received surgical treatment (n = 45)

	AIS grade after two years				
Initial AIS grade	A	B	C	D	E
A	9				
B	1	3	3	3	
C	1		4	7	1
D	1			2	10
AIS, American Spinal Injury Association (ASIA) Impairment Scale					
The *P* value of the McNemar test was 0.004					
Patients who died were included into “A” group at AIS grade during follow-up period.					

Table 4
Baseline characteristics in patients with ankylosing spondylitis according to spinal cord injury or spinal fracture

	Spinal cord injury			**Spinal fracture**		
Variables	SCI (*n* = 60)	Non-SCI (*n* = 45)	*P* value	Fracture (*n* = 94)	Non-fracture (*n* = 11)	*P* value
Subluxation or dislocation	29 (48.3)	4 (8.9)	< 0.001	32 (34.0)	1 (9.1)	0.167
Location of spinal fractures			< 0.001			-
None	11 (18.3)[a]	0 (0.0)		-	-	
C0-2	1 (1.7)	4 (8.9)		-	-	
C3-7	32 (53.3)[a]	5 (11.1)		-	-	
T	8 (13.3)[a]	20 (44.4)		-	-	
L	1 (1.7)[a]	14 (31.1)		-	-	
Multiple regions involved	7 (11.7)	2 (4.4)		-	-	
Spinal cord injury level			-			0.001
None	-	-		45 (47.9)[a]	0 (0.0)	
C1-2	-	-		3 (3.2)	0 (0.0)	
C3-7	-	-		35 (37.2)[a]	11 (100.0)	
T	-	-		11 (11.7)	0 (0.0)	
Bamboo spine	47 (78.3)	31 (68.9)	0.367	74 (78.7)	4 (36.4)	0.006

SCI, spinal cord injury; SEH, spinal epidural hematoma

Values are given as number (%) or mean ± standard deviation

‘a’ indicates the value reached a significant difference between the two proportions in the row

	Spinal cord injury			Spinal fracture		
SEH	13 (21.7)	1 (2.2)	0.003	14 (14.9)	0 (0.0)	0.353
SCI, spinal cord injury; SEH, spinal epidural hematoma						
Values are given as number (%) or mean ± standard deviation						
‘a’ indicates the value reached a significant difference between the two proportions in the row						